

Zoonotic Anatrachosomiasis in a Mother and Daughter

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Zoonotic anatrachosomiasis in a mother and daughter is reported. Both presented with a 10-week history of multiple painful oral ulcers. Biopsy specimens revealed the presence of small, coiled trichuroid nematodes with distinctive morphological features, including stichocytes and paired bacillary bands. This represents an unusual infection by a zoonotic *Anatrachosoma* species.

CASE REPORT

The anatrachosomes are an unusual and poorly known group of trichuroids that have been reported from primates, marsupials, and, rarely, humans, dogs, and cats. The adult worms are small and delicate, and most often they have been reported burrowing in epithelial tissues of skin or mouth or from cutaneous lesions. Between 1960 and 1990, there were three reports of infection in humans with *Anatrachosoma*, all from cutaneous trails in the skin similar to cutaneous larva migrans (1–3), and in 2005, a report of an *Anatrachosoma*-like worm was reported in a breast nodule (4). More recently, a single case from the United States was reported in which the worms were recovered from lesions in the mouth (5).

Here we report our findings on two patients, a mother and daughter, in whom we identified zoonotic *Anatrachosoma* infection. This is the second report worldwide of anatrachosomiasis involving the mouth.

Two women, a mother and daughter, ages 52 and 29, respectively, presented with painful lesions on the tongue, lips, hard and soft palate, and buccal mucosa (Fig. 1) of approximately 10 weeks duration. Individual lesions were transitory and lasted 3 to 5 days each. Lesions tended to occur in “crops.” Both women lived in the greater metropolitan Iowa City area. Symptoms developed shortly after returning from a 2-week trip to Puerto Vallarta, Mexico, in August 2013; there was no other recent travel history outside the United States. Of note, but uncertain relevance, was the fact that the mother and daughter who experienced the oral lesions had participated in what are termed “snail facials.” Although available at their hotel’s spa, they performed their facials themselves by collecting native snails obtained in the tidal region of the rocky shore. The rocky shore, besides having snails, also exhibited bird guano, although they attempted to avoid the guano. The snails were placed live on the face and allowed to crawl on the skin of the cheeks and forehead (as is done in typical “snail facials”). Snails were not allowed to come in contact with the lips or mouth. For more information and photos of snail facials, a simple internet search of “snail facial” will work. A second daughter who was also on the trip but did not have any symptoms, ate all the same foods and traveled to all locations during their entire vacation but did not participate in the snail facials or have contact with any of the snails. The two daughters deny having much or any outside activities in Iowa, although the mother does some light yard work. Their residence in Iowa is on several acres, and they have noticed opossums and raccoons on their property but deny any close contact.

Medical history reported that both patients had been seen pre-

viously by several health care providers and had been treated with several different antibiotics (ciprofloxacin, amoxicillin/clavulanate potassium) and antivirals (valacyclovir) for the condition, as well as a methylprednisolone (Medrol; Sandoz, Inc., Princeton, NJ) dose pack (which is a tapered regimen of 24 mg on day 1, 20 mg on day 2, 16 mg on day 3, 12 mg on day 4, 8 mg on day 5, and 4 mg on day 6 of methylprednisolone) and a dexamethasone 0.5 mg/ml elixir as a rinse. The patients reported no amelioration of symptoms with any of these therapies, and the lesions worsened with methylprednisolone as well as dexamethasone treatments.

At examination, the both patients had multiple areas of ulceration, approximately 1 to 2 mm wide but extending as much as a centimeter to centimeter and a half. One particular lesion on the left lateral dorsal surface of the daughter’s tongue appeared to be indurated. The areas of involvement at that time appeared to be the soft palate, tongue, inner lip, buccal mucosa, and the palatal gingiva. No lesions were observed or reported by the patients to occur on the outer lip region or any area of the facial skin.

The lesion on the lateral tongue of the daughter was biopsied on 2 separate occasions, 1 week apart, and submitted for routine histologic sectioning. Microscopic examination of the hematoxylin and eosin (H&E)-stained sections revealed the presence of small, coiled nematodes lying superficially in the mucosa (Fig. 2). Closer examination of the worms revealed morphological features compatible with *Anatrachosoma* spp., including a thin cuticle that was thicker over the lateral chords, a diameter of no more than 40 μ m, triradiate esophagus, paired lateral bacillary bands, and a very prominent stichosome composed of many stichocytes in the anterior end. All sections available for study were through the anterior portions of the worm, so the maximum diameter could not be determined, and no sections contained reproductive structures. The biopsy specimen taken from the mother failed to show evidence of the organism.

Buccal mucosa scrapings with a small brush were taken to see if either eggs or adult worms might be recovered to further facilitate

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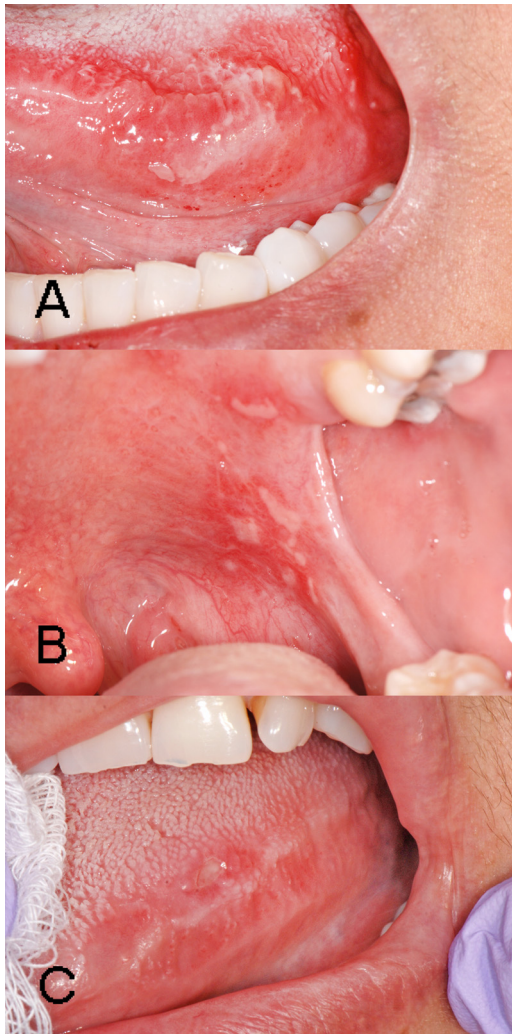


FIG 1 Clinical images showing lesions in the buccal cavity. (A) Large ulcerated area on the lateral margin of the daughter's tongue that was the site of the first biopsy. (B) Raised white plaques and track-like lesions on the palate of the daughter at the time of first biopsy. (C) Ulcerated lesion on the mother's tongue that was the site of biopsy.

identification. No parasite material was observed in any of the scrapings.

A tissue block remaining after much sectioning and which possibly contained a very small bit of worm was submitted to the Infectious Disease Pathology Branch, CDC, for DNA extraction. Unfortunately, only human DNA could be amplified.

Both patients were treated with 400 mg of albendazole once daily for 3 days, and a positive therapeutic outcome was achieved in both. All oral lesions resolved with no recurrence of symptoms over the course of a 4-month follow-up.

This is only the second time that *Anatrichosoma* has been reported from the buccal cavity of humans. In the first case, a Hispanic man living in Illinois but with recent travel to several points in Mexico presented with very similar symptoms and oral lesions observed in the present cases (5). That the two present cases also had recent travel to Mexico is highly suggestive that infection in all

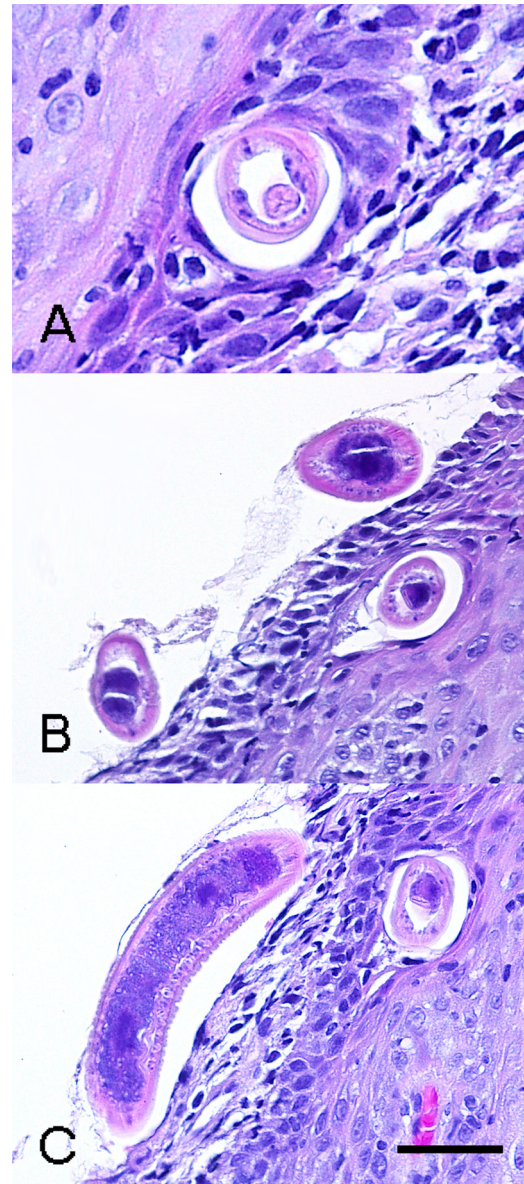


FIG 2 Photomicrographs of histologic sections of the parasite seen in the biopsy specimen from the mother. (A) Cross section of the anterior end of the worm (anterior to the stichosome region) through the region of the esophagus with triradiate lumen. Scale bar, 25 μ m. The lateral bacillary bands are evident (at the 2:00 and 8:00 o'clock positions). Note the unevenly thick cuticle. (B) View through multiple sections of the worm showing the presence of darkly staining stichocytes in all three sections. Scale bar, 50 μ m. (C) Tangential section (to left) illustrating the stichosome composed of numerous stichocytes. Scale bar, 50 μ m.

three cases was acquired in Mexico. However, it cannot be discounted that all three infections may have been acquired in the Midwest. It may also be coincidental that the two present cases engaged in snail facials. As has been previously noted, the life cycle for *Anatrichosoma* is unknown but thought to be direct (6). The eggs which contain a larva are sloughed and passed out of the body of the host and may be infective to the final host at that time. However, some related genera in the subfamily Capillariinae require an insect intermediate host for further development before

becoming infective to the definitive host. There was nothing unusual in the dietary history of either of the present cases or the previous case that suggested a possible source of exposure.

There are several recognized species of *Anatrichosoma*: one or more from the nasal mucosa of primates in Southeast Asia and Africa, one from the eye of tree shrews in Southeast Asia, one from gastric mucosa in gerbils in Tunisia, one from paracloacal glands in marsupials from Australia, and one from the buccal mucosa of opossums in the Americas (6). The species in opossums, *Anatrichosoma buccalis*, is the most likely cause of the infections reported here in humans based both on known geographic distribution in the Americas and location within the host (5, 7). However, no distinctive morphological features have been described in tissue sections that could be used to identify or distinguish between species. Furthermore, at present there is very limited genetic typing of any *Anatrichosoma* species available, so molecular comparisons await further work, as will a more definitive diagnosis of the species involved.

There also have been reports of anatrachosomiasis in skin lesions in dogs, cats, monkeys, and humans, but none of these have been ascribed to a specific species, and the relationship between the worms in skin and those in the buccal cavity or nasal mucosa is not understood.

The differential diagnosis included *Gongylonema*, a common nematode in domestic and wild animals and birds that is known to occur on rare occasions in the mucosa and submucosa of the mouth and tongue in humans (8, 9); approximately 13 cases have been reported in the United States (8). In contrast to cases of anatrachosomiasis, *Gongylonema* worms are much larger, and the presence of the worm is usually noted by visual examination, either by the patient themselves or by health care providers. In addition to the greater size, the presence of distinctive cuticular bosses on the anterior end of *Gongylonema* further differentiate it from *Anatrichosoma*, as do the trichuroid features present in *Anatrichosoma* noted above, most notably the stichocytes and sti-

chosome, which are readily evident in tissue sections. *Capillaria*, another trichuroid group, was also considered because members of that genus frequently inhabit tissues. However, *Capillaria* worms typically have three bacillary bands and do not have thickened cuticle in the anterior end as was present in this case.

We have now been able to document successful treatment of three cases with the first treated with mebendazole for 20 days and the 2 recent cases with albendazole for 3 days. In all three patients, the course of treatment was well tolerated, though the albendazole provided more rapid amelioration and no recurrence of symptoms.

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